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Late recurrence of stage Ia ovarian mucinous cystadenocarcinoma in teenage patients

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ABSTRACT

We present the case of a 13-year-old patient who underwent right salpingo-oophorectomy and was diagnosed with stage Ia ovarian mucinous cystadenocarcinoma. Nine years after complete resection, she was diagnosed with metastasis of the previously resected carcinoma. Despite receiving combination chemotherapy, the patient died of the disease. Long-term follow-up of teenage patients with stage Ia ovarian mucinous cystadenocarcinoma is important.

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Epithelial ovarian carcinoma (EOC) is the leading cause of death due to gynecological malignancies [1]. The standard surgical treatment for patients with EOC involves hysterectomy and bilateral salpingo-oophorectomy with peritoneal sampling (peritoneal washing, omentectomy, multiple peritoneal biopsies, and peritoneal implant removal), with or without lymph node sampling [2]. Several reports have estimated that 3%–17% of all EOCs develop in women under 40 years of age and that the incidence of this condition in teenage patients is rare [3]. Current studies on patients with stage I EOC suggest a comparatively more favorable prognosis on treatment with FSS compared radical surgery [2–6]. It has also been shown that the long-term outcome of patients with stage Ia mucinous carcinomas with no apparent disease in the peritoneum is favorable [7]. Herein, we report the case of a teenage girl with late recurrence who was previously treated for stage Ia ovarian mucinous cystadenocarcinoma. We also performed a literature review of case reports describing recurrence after tumor resection in teenagers with stage Ia EOC.

1. Case report

A 13-year-old girl was referred to our hospital because of abdominal distension. Her family history was unremarkable. Physical examination and subsequent abdominal ultrasonography and computed tomography (CT) confirmed the presence of a very large solid cystic mass in the lower abdomen (Fig. 1). Serum levels of carcinoembryonic antigen (CEA, 16.2 ng/mL; normal range, below 10 ng/mL), carbohydrate antigen 19-9 (CA19-9, 2249.0 U/mL; normal range, below 37 U/mL), and carbohydrate antigen 125 (CA125, 49.9 U/mL; normal range, below 40 U/mL) were considerably elevated. The patient underwent right salpingo-oophorectomy, and a right ovarian tumor was diagnosed. Visualization and palpation did not reveal disease. Wedge resection of the remaining ovary was not performed. Pathological examination of the right ovary revealed multiloculated cysts lined by tall mucin-secreting columnar epithelial cells. In addition, at places with epithelium stratification, papillary and cribriform patterns were observed. The pathological findings of the washings were negative. International Federation of Gynecology and Obstetrics stage Ia (T1aN0M0), grade 1 ovarian mucinous cystadenocarcinoma was diagnosed. After surgery, the levels of each tumor marker returned to normal within 3 weeks. The patient was discharged from the hospital after experiencing complete remission, and she was followed closely without adjuvant therapy.

Tumor marker levels were examined twice every year. Serum levels of CEA, CA19-9, and CA125 were low during the first 7 years after the operation. Nine years after complete tumor resection (21 years old, gravida 0, para 0), the patient complained of respiratory discomfort. Chest radiography and CT confirmed the presence of multiple coin lesions in the bilateral lung fields. Laboratory investigation of tumor marker levels (CEA, 24.6 ng/mL; CA19-9,

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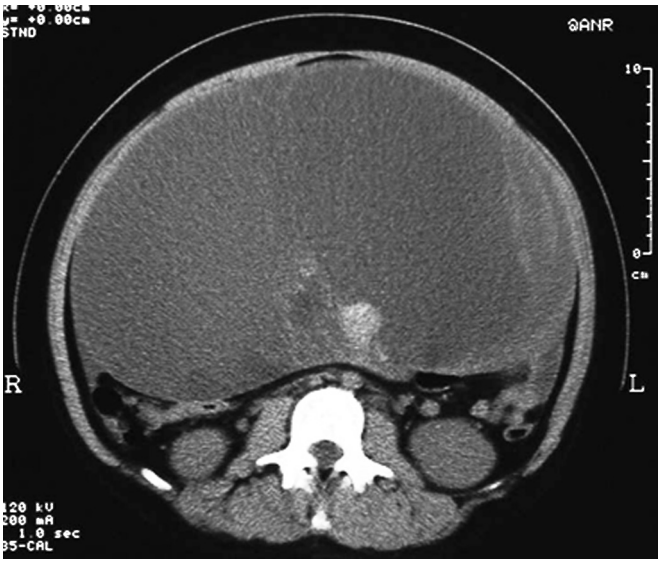

Fig. 1. Computed tomography (CT) scans confirmed the presence of a very large solid cystic mass in the lower abdomen.

9860.0 U/mL; and CA125, 245.8 U/mL) revealed that they were elevated. Bronchoscopy revealed that the superior segment was incompletely occluded by angiectasis and adenomatous hyperplastic epithelium. Transbronchial lung biopsy revealed atypical gland formation the lung tissue, including in the alveoli. Expression of thyroid transcription factor-1 was negative. The biopsy findings suggested that metastasis of the previously resected ovarian mucinous cystadenocarcinoma had occurred. Fluorine-18 fluorodeoxyglucose positron emission tomography/CT revealed metastasis to the left lung, pulmonary hilum, left buccal region, liver, mediastinal space, pelvic space, bilateral arms, and subcutaneous layers. Bone scintigraphy revealed hot lesions at the pelvic bone, bilateral femoral bone, skull bone, spine, and costal bone. Recurrent ovarian mucinous cystadenocarcinoma was diagnosed, and the patient was treated with carboplatin and paclitaxel combination chemotherapy. The chemotherapy regimen comprised of 794 mg carboplatin (area under the curve of 4.5) and 237 mg paclitaxel (170 mg/m^2). Despite the combination chemotherapy treatment, levels of each tumor marker increased, and the patient died of the disease 5 months after the diagnosis of recurrence.

2. Discussion

The key to feritility-sparing surgery (FSS) is to have adequate information about the biology and stage of the cancer. FSS for patients of reproductive age with invasive EOC has been adopted for stage Ia tumors according to the 2007 guidelines of the American College of Obstetrics and Gynecology [8] and for unilateral stage I tumors without dense adhesions and with favorable histology according to the 2008 guidelines of the European Society for Medical Oncology [9]. In Japan, FSS has been recommended for patients with stage Ia tumors [10].

Characteristics of recurrence after FSS for stage Ia EOC in teenagers as described in the literature are shown in Table 1 [2,4–6]. Recurrence after tumor resection has been described in 8 of these patients, including the patient described here (Table 1). The age of the patients at the initial diagnosis ranged from 14 to 19 years (average age, 16.8 years). The histological findings of these cases indicated low-grade mucinous cystadenocarcinoma. The size of the primary tumor could be determined for 3 of the 8 patients and ranged from 30 to 50 cm (median, 40 cm). None of the 8 patients received adjuvant chemotherapy. Of the 8 patients, 4 had relapses involving the remaining ovary and 3 had relapses involving the lungs. No patients died of local recurrence. The recurrence-free interval was 2–118 months (median, 74 months). Four of 6 patients died of disease within 5 years of recurrence being diagnosed. In the treatment of recurrence, salvage surgery of contralateral salpingo-oophorectomy may not be effective, and distant recurrence sites were refractory to chemotherapy. Recurrence occurs late and is systemic in teenage patients with stage Ia EOC. Cases of large mucinous cystadenocarcinomas in teenagers are characterized by a poor prognosis. Chan et al. [11] reported that age, stage, grade, and cytology are important prognostic factors for high-risk early-stage EOC. Although these data are based on a very small amount of retrospective data, the strategy of surgical treatment and adjuvant therapy chosen are important for teenage patients with early-stage EOC. After surgical resection, selecting a treatment strategy based on the biology of each individual's tumor is also important.

Adjuvant chemotherapy is associated with improved recurrence-free survival in patients with early-stage ovarian cancer with

Table 1
Case reports of recurrence after tumor resection for stage Ia epithelial ovarian carcinoma in teenage patients.

Ref.	Age at diagnosis (years)	Histology	Grade	Size of the tumor (cm)	Rupture	Surgical procedure	Adjuvant chemotherapy	Site of recurrence	Recurrence-free interval (months)	Treatment for recurrence	Follow-up interval after recurrence (months)	Status
Satoh et al. [8]	18	Muc	1	NA	NA	USO	No	Remaining ovary	83	Salvage surgery	119	NED
	19	Muc	1	NA	NA	USO	No	Peritoneum	70	NA	149	NED
Park et al. [9]	15	Muc	NA	50	No	USO	No	Remaining ovary	41	Ovarian cystectomy	NA	NED
	15	Muc	NA	30	No	USO	No	Lung, pericardium	82	Biopsy	NA	DOD
Morice et al. [2]	18	Muc	1	NA	NA	USO	No	Remaining ovary	7	Surgery, ChT	40	DOD
	16	Muc	2	NA	NA	USO	No	Remaining ovary	2	Surgery	54	DOD
Schilder et al. [10]	19	Muc	2	NA	NA	USO	No	Lung	78	RT, ChT	19	DOD
Present case	14	Muc	1	40	No	USO	No	Lungs, bones	118	ChT	5	DOD

EOC, epithelial ovarian carcinoma; Muc, mucinous cystadenocarcinoma; USO, unilateral salpingo-oophorectomy; ChT, chemotherapy; RT, radiation therapy; NED, no evidence of disease; DOD, died of disease; NA, not available.

nonoptimal staging [12]. However, in teenage patients with recurrent early-stage EOC treated with unilateral salpingo-oophorectomy without adjuvant therapy, long-term prognosis is not as favorable as that described in adult patients. This is true even for patients with stage Ia tumors [2–6]. We should be aware of the possibility of recurrence in teenage patients with primary large mucinous cystadenocarcinoma.

3. Conclusion

Herein, we report a case of late recurrence in a teenage girl previously treated for stage Ia ovarian mucinous cystadenocarcinoma. Long-term follow-up of teenage patients with stage Ia ovarian mucinous cystadenocarcinoma is therefore important.

Conflicts of interest

The authors have no conflicts of interest to report.

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